Behavioral Approaches To The Treatment Of Sleep Problems In Children with Developmental Disorders: What Is The State Of The Art?

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This paper reviews behavioral treatments for sleep problems in children with a developmental disorder (DD). Sleep problems are common in children with a DD and children's sleep problems may be associated with adverse consequences including behaviour problems, compromised daytime functioning and family stress. However, the sleep intervention literature for these children is seriously lacking, with only extinction and graduated extinction approaches meeting criteria for a probably efficacious treatment for common sleeplessness problems. The investigation of behavioral treatments for other sleep difficulties remains in its infancy. The impact of sleep problems and successful treatment is largely unexplored, while professional awareness regarding sleep problems and their treatment appears poor.
Key Words: Sleep – developmental disorders – sleeplessness - parasomnias – excessive sleepiness - behavioural treatment

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Sleep disturbances are one of the most frequently reported behaviour problems affecting children from the general population, with estimated prevalence rates of about 30% (Richman, 1981; Owens, Spirito, McGuinn, & Nobile, 2000). For children with developmental disorders (DDs), sleep disturbance is even more widespread and problems are typically more severe and persistent. Amongst children with intellectual disabilities (IDs) up to 86% of children aged less than six years were reported by parents to have 'sleep problems' (Bartlett, Rooney, & Spedding, 1985) with similar high rates (77%) even for children aged 12-16 years. Problems of sleeplessness (i.e., difficulty getting off to sleep, night waking, or early waking) appear to be the most commonly reported problems (Quine, 1991; Wiggs & Stores, 1996a). Prevalence data for aetiologically discrete samples is limited but studies have suggested that between 44% and 89% of children with autistic spectrum disorders (ASD) may have sleep difficulties, again with sleeplessness featuring prominently (Richdale, 1999; Wiggs & Stores, 2004). Interestingly, amongst ASD samples, high rates of sleep problems appear to occur independently of the presence of co-existing ID (Richdale & Prior, 1995; Patzold, Richdale & Tonge, 1998). True prevalence rates are difficult to assess and likely to vary depending on the age and nature of the group of children studied and the types of sleep disturbances investigated but it seems clear that perhaps the majority of children with DDs suffer from some form of sleep disturbance.

There are over 80 different sleep disorders listed in the International Classification of Sleep Disorders (American Sleep Disorders Association [ASDA], 2001) but there are three main types of presenting sleep problems: sleeplessness, excessive sleepiness and episodes which are associated with/intrude into sleep (i.e., parasomnias). Most of the literature concerning sleep patterns of children with DDs does not make the distinction between 'sleep problems' and 'sleep disorders', with research largely describing presenting symptoms (e.g., "difficulty getting off to sleep") based upon parents report. This is a limitation because management decisions need to be based upon correcting the underlying disorder and different sleep disorders can present with similar symptoms. For example, sleeplessness, taking the form of a difficulty getting to sleep at night, may amongst other things, result from disorders of the body clock, failure to learn appropriate bedtime behaviour or anxiety. Treatment for each of these causes, or underlying disorders, would be very different.

The Significance of Sleep Disturbance

The widespread problem of persistent sleep disturbance in children with DDs is especially concerning in view of the fact that sleep disturbance is associated with far-reaching and serious effects (Stores, 1996; 2001; Pilcher & Huffcutt, 1996; Fallone, Owens, & Deane 2002), likely to further compromise daytime functioning in children whose daytime functioning is already impaired by virtue

of their underlying condition. Effects of sleep disturbance include cognitive deficits, perhaps especially impaired attention, memory and creative thought, measurable decrements in academic performance at school, behavioral problems, increased reports of depressed mood and irritability (Sadeh, Gruber, & Raviv, 2002) and even physical factors such as impaired growth and immune function (Pollmacher, Mullington, Korth, & Hinze-Selch, 1995). For children, who typically live within a family unit, their sleep disturbance often also adversely affects the sleep patterns and thus subsequent daytime functioning, of other family members. Further, an association between childhood sleep problems and general family stress, marital discord and parenting difficulties has been found (Quine, 1991; Richdale, Gavidia-Payne, Francis, & Cotton, 2000). Of note is that many of the adverse effects can be reversed by removal of the sleep disturbance, emphasizing the importance of addressing sleep disturbance as a means of perhaps improving overall functioning of the child and family unit.

Some recent experimental work with typically developing (TD) children who were 'good-sleepers' suggests that the amount of sleep loss needed to induce impairments in neuropsychological functioning is quite subtle (i.e., a matter of about 30 minutes). And conversely, that extending normal sleep by a similar small amount can lead to improvements (Sadeh, Gruber, & Raviv, 2003). As such, efforts should be made to ensure that children are receiving adequate, good quality sleep sufficient to meet their individual sleep needs and perform optimally. There will be intra- and inter-individual differences in sleep needs, depending on a variety of factors both internal and external to the individual (Roehrs, Carskadon, Dement & Roth 2000). Determining whether an individual has impaired sleep quality or quantity is best achieved by careful examination of sleep timing, sleep-related behaviours, sleep physiology, daytime sleepiness/functioning and possibly by experimental behavioural manipulation of sleep times (Stores 2001).

Assessment of Sleep Patterns

It is beyond the scope of this review to provide detailed information about assessment options and procedures and readers are directed to look at Stores (2001) and Mindell and Owens (2003) where such information is provided. The importance of careful clinical enquiries in the form of a sleep history to assess the precise nature and course of the problem along with a developmental, medical, psychological and family history is emphasised. Reviewing the child's 24 hour sleep wake pattern in some detail is also recommended as a method of identifying factors which are causing or maintaining sleep problems. Where appropriate, further additional enquiries may be made. Stores (2001) describes the various options in detail which include the use of sleep diaries in which parents can systematically record information, questionnaires (a variety exist both for screening purposes and to provide detailed information about specific aspects of sleep for devising and monitoring treatment) and more objective methods such as video recordings, monitoring of body movements by means of actigraphy (which can detect basic sleep wake patterns) and conducting polysomnography (PSG), either in a laboratory or in the child's home, to record various physiological parameters which permit detailed analysis of sleep physiology according to standardized criteria. Objective recordings may be helpful in cases of diagnostic uncertainty, where the clinical information is lacking or doubtful in its veracity and also for assessment of sleep disorders where features of the PSG form part of the formal diagnostic requirements (e.g., narcolepsy, obstructive sleep apnoea). However, the need for objective recordings should not be overemphasised. Clinical enquiries and systematic sleep diary information will, in many instances, form the essential basis of assessment and a means of monitoring treatment.

There are a variety of factors, both physical and psycho-social, which might combine to predispose children with DDs to elevated rates of sleep disturbance and which need to be considered in relation to assessment. Firstly, brain maldevelopments or damage have been seen to be associated with abnormalities of sleep physiology and sleep-wake patterns, depending on the extent and site of brain abnormality (Okawa & Sasaki, 1987). Also, the child's underlying disorder may be associated with particular physiological abnormalities that predispose to certain types of sleep disorders (e.g., the

congenital narrowing of airways, reduced muscle tone and enlarged tongues of children with Down syndrome predisposing to sleep-disordered breathing difficulties) (Stores & Wiggs, 2001).

Secondly, medical issues feature prominently (e.g., craniofacial abnormalities, obesity, seizure disorders, muscle disease, medication use) and may in themselves directly lead to sleep disturbance. Sensory impairments are also more likely and sleep problems have been seen to be prominent for both children with visual impairment (attributed to the effects of impaired/no light perception on melatonin production and reduced social/environmental cues) and those with hearing impairment (reduced social/environmental cues plus possible effects of tinnitus, anxiety associated with hearing-aid removal at night). Medical and physical factors may, of course, increase the likelihood of salient sleep-related psychological issues (e.g., anxiety) in the parent and/or child and where both physiological and psychological factors contribute to sleep disturbance both causes may need to be addressed.

Thirdly, and importantly, elements of 'sleep' are learned, including aspects of when and how to fall asleep. Children with IDs may find it particularly difficult to learn appropriate behaviours (or to unlearn inappropriate behaviours). Quine (1991) suggests that communication problems are predictive of the development of sleep problems in young people with severe IDs, highlighting one possible reason for why it may be difficult to teach necessary behaviours. It may similarly be more difficult to establish good sleep habits in children who are emotionally or behaviorally disturbed and of course such problems are encountered in intellectually impaired children more than in other children (Borthwick-Duffy, 1994).

Fourthly, psychological parental factors may play a role. Stress, marital problems, mental health problems and anxiety about the child, all of which can be elevated amongst parents of children with DDs (Quine & Pahl, 1985) may make it more difficult for parents to develop and maintain a consistent, disciplined approach to their child's sleep pattern and to instil independent sleeping habits.

Lastly, there is frequently no attempt made to treat the sleep disturbances of children with DDs. Both parents and professionals may be unaware that many sleep problems can be successfully treated, even if the problems are severe, long-standing and complicated by physical and psychological aspects of the child's basic underlying condition. A significant number of these children do not receive treatment for their sleep problem (Robinson & Richdale, 2004; Wiggs & Stores, 1996a). Medical practitioners may not enquire about or recognise sleep problems in children and / or parents may fail to raise this issue. For example Chervin et. al. (2001) found that US pediatricians discussed sleep problems in only 16 of 103 cases over two years, treating only three of these; Owens (2001) also found that there were significant gaps in US pediatricians' knowledge about and treatment of sleep problems in children and adolescents; and most recently Blunden et al. (2004) found that Australian, school-age children's sleep problems were considerably under-reported by parents during general practice consultations.

Children with DDs may be particularly overlooked by professionals, as their parents often do not seek help (Robinson & Richdale, 2004; Wiggs & Stores, 1996a). Reasons for this appear to be wide ranging but can include habituating to the difficulties if they are long-standing, erroneously viewing them as inevitable or untreatable and even unawareness of the problem (the latter perhaps particularly likely with sleep related breathing disorders). Studies which have focussed on those who have received treatment for sleeplessness suggest that medication commonly was prescribed though its effectiveness was poorly rated by parents (Wiggs & Stores, 1996a), while Robinson and Richdale (2004) found that parents often rated treatments poorly, regardless of their type. Taken together, research suggests it is important to routinely screen for the presence of sleeping difficulties in order to avoid the harmful effects of sleep disorders on the child and their family and for clinicians' education about sleep to be improved upon so that they are able to implement appropriate and effective forms of treatment.

Treatment of Sleep Disorders

Because the term 'sleep disorder' encompasses such a broad range of disorders there is a range of approaches to the treatment of sleep disorders, including basic explanation of environmental and lifestyle factors that can affect sleep; behavioral techniques, which aim to teach the child appropriate sleep habits and/or to correct inappropriately learnt behaviour; cognitive therapy; chronotherapy (altering sleep timing in specific, systematic ways to re-set the body clock); physical measures such as using bright light to alter sleep times; or using aids such as nasal continuous positive airways pressure (NCPAP) to aid breathing during sleep; surgery (e.g., removal of tonsils and adenoids if they are causing an obstruction); and pharmacological approaches (e.g., sedatives, stimulants, melatonin). Choice of which treatment to use is of course, in part, dictated by the underlying cause of the sleep disorder. In general, there is a need for considerably more research to establish the efficacy and relative efficacy of various management options with children with specific types of sleep disorders and discrete basic underlying conditions.

Evidence suggests that medications commonly are prescribed as a treatment for children's sleep problems in the USA (Mindell & Owens, 2003; Owens, Rosen, & Mindell, 2003), the UK (Bramble, 2003) and in Australia (Efron et al., 2003). Pharmacological approaches may have a role to play in the treatment of some sleep disorders, for example, clonidine and impramine have been reported as useful in children with ADHD and melatonin has been found to be helpful for some children with disorders of the sleep-wake cycle, although the uncertainties and concerns surrounding the use of melatonin in children must be carefully considered in each individual case (Stores, 2003). The use of sedative-hypnotic medications generally lack empirical support and has a very limited role to play in the management of children's sleeplessness since the clinical effects are slight and not well maintained when compared to behavioral interventions (Ramchandani, Wiggs, Webb, & Stores, 2000). In addition, hangover daytime effects, paradoxical responses and parental resistance to its use are common (Kuhn & Weidinger, 2000). Owens et al. (2005) provide helpful consensus guidelines for the use of pharmacotherapy in the treatment of pediatric insomnia (including particular considerations with children with DDs and other special needs) stressing the need for pharmacotherapy, where indicated, to be used in conjunction with behavioral therapy.

Behavioral interventions have been used to successfully treat a variety of different sleep disorders and are the treatment of choice for many of the most common pediatric sleep disorders. They are particularly appropriate for interventions with children with ID since they utilize non-verbal methods of modifying behaviour. Further, they can be individually tailored to each child and family's particular needs and circumstances, which is especially important when dealing with children with complex and multiple problems. Of further note is that their successful use has been associated with improvements in child behaviour and various aspects of parental functioning (e.g., Quine, 1992; Minde, Faucon, & Falkner 1994).

This paper will review the literature on the use of behavioral therapy in the treatment of sleep disturbance in children with DDs. It is important to note that only behavioral techniques which have been subject to empirical research with this population are included in the current review and it is therefore not a comprehensive review of all the behavioral interventions which might ultimately be considered helpful for treating sleep disorders in children with DDs.

Sleeplessness in Children with Developmental Disorders (DDs)

Disorders of sleeplessness (dyssomnias) are probably the most common sleep complaints of parents of young children and parents of children with DDs. Sleeplessness in children most commonly includes settling and bedtime refusal, co-sleeping and night waking in both TD children and children with DDs, particularly under age 5 years. For example, Cotton and Richdale (in press) found that these were the most common complaints reported by parents of young and school age children with autism, Down syndrome or global developmental delay. Common causes of

sleeplessness in children are behavioral and/or psychological in nature, for example a failure to set limits on the child's behaviour may result in bedtime refusal, while night time fears or anxieties may prevent a child from easily falling asleep.

The psychological treatment of settling and night waking problems in young TD children has been evaluated against definitions for empirically validated psychological interventions (Chambless & Ollendick, 2001; Lonigan, Elbert, & Johnson, 1998) and both parent education about children's sleep and extinction have been shown to be well established treatments (Mindell, 1999). Similarly Kuhn and Elliot (2003) concluded that behavioral interventions were empirically supported for bedtime refusal and night waking in children, noting the lack of empirically supported interventions for other infant and child sleep problems. Schreck (2001) reported that extinction was a probably efficacious treatment for settling and night waking in children with autism. However, in general adequate evaluations of sleep interventions for children, and in particular children with DDs, are lacking and advice is based on clinical experience, limited numbers of studies (generally case studies), and the adult literature (Glaze, Rosen, & Owens, 2002). However encouragingly, there are an increasing number of behaviorally based treatments for sleeplessness in developmentally delayed children now being published (see Table 1). A review of studies addressing sleeplessness in children with DDs and using a behavioral approach follows.

Table 1. *Behavioural Interventions for Sleeplessness*

Study	Intervention	Design	*N*	Disability	Age (years)	Sleeplessness	Success		
							Post Intervention	Follow-up	Follow-up Success
Bramble (1997)	Routine & Extinction	AB cases analysed as a group data	15	Severe ID, 3 with DS, range of other disabilities	3.5 - 12	Settling, night waking, co-sleeping	Yes	4 & 18 months	Yes
Christodulu & Durand (2004)	Routines & Sleep restriction	Multiple baseline	4	Dev delay with CHARGE association, PDDNOS, IGA deficiency and ankyoglosia, autism	2.5 - 5.9	Settling, night waking, co-sleeping	Yes Routine only worked for 1 child	1 month	Yes
Curfs, Didden, Sikkema, & De Die-Smulders (1999)	Routine & extinction	Functional assessment, case study	1	Severe ID, Wolf-Hirschhorn syndrome	6	Settling, night waking	Yes	6 months	Yes
Didden Curfs, Sikkema, & de Moor (1998)	Routine, Extinction	Functional assessment, case study series	6	Severe ID, PWS	4	Settling	Yes	6 months	Yes
				Mod ID, spastic diplegia, strabismus	2	Settling, night waking, co-sleeping	Yes	3 months	Yes
				Mild ID, ADHD	6	Settling, co-sleeping	Yes	6months	Yes
				Mild ID, spinal muscular atrophy	2	Settling, co-sleeping, night waking	Yes	3 months	Yes,
	Treatment for epilepsy, Extinction			Mod ID, Spastic tetraplegia, epilepsy, asthma	2	Settling, night waking	No	3 months	No

	Desensitisation, differential reinforcement			Mild ID, FRX	7	Settling, co-sleeping	Yes	6 months	Yes
Didden, Curfs, van Driel, & de Moor (2002)	Routine & extinction	Functional assessment, non-concurrent multiple baseline	4	Severe ID, epilepsy, visual & physical impairment	7.25	Night waking	Yes	6 months	Yes
				Mod-severe ID, autism, epilepsy	6.4	Settling, night waking	Yes		Yes
				Severe ID, DS	25	Settling, night waking, co-sleeping	Yes		Yes
				Mild Dev delay, lang. delay, motor delay	1.9	Settling, night waking	Yes		Yes
Didden, de Moor, & Curfs (2004)	Routine, Extinction	Functional assessment, non-concurrent multiple baseline	3	Mod ID, DS	9.2	Settling, co-sleeping;	Yes	6 months	Yes
				Severe ID, epilepsy,	12.4	night waking;	Yes		Yes
	DRI + response cost + tokens + punishment			Mild ID / ADHD	10	night waking	Yes		Yes
Didden, de Moor, & Kruit (1999)	Routine, Extinction	Functional assessment, case study	1	Mild ID, leftside paresis	2.4	Night waking	Yes	3 months	Yes
Durand & Christodulu (2004)	Sleep restriction	Multiple baseline	2	Dev delay & autism	4	Settling, night waking, co-sleeping	Yes		None
				Developmental delay	4	Settling, night waking. Co-sleeping	Yes		None
Durand Gernert-Dott, & Mapstone (1996)	Routine, Graduated extinction	Functional assessment, multiple baseline, 2 groups	4	Mild-mod ID, DS,	11	Night waking	Yes	None	-
				Mild-mod ID, 9th monosme syndrome	7	Night waking, co-sleeping	Yes	6 months	Yes
				Dev delay, visual impairment	2	Settling	Yes	None	-
				Severe ID, autism	12	Settling	Yes	6 months	Yes
Hewitt (1985)	Routine, Relaxtion Stimulus fading	Functional assessment Case study series	10	Severe ID	3.2	Night waking	Yes	12 months	Yes
				DS	3.8	Settling	Yes	5	Yes
						Night waking	Yes	months	Yes
				DS	4.25	Settling	Yes	6	No
						Night waking	Yes	months	No
				DS	4.4	Night waking Head banging	No	None	-
				DS	5.8	Night waking	Yes	12 months	Yes
				DS	6.2	Settling	Yes	12 months	Yes
				DS	6.5	Settling	Yes	12 months	Yes
				DS	16.5	Night waking	Yes	12 months	Yes
				Cornelia de Lange sydnrome	8.6	Night waking	Yes	1 month	No
								12 months	No
				Tuberous sclerosis	12.5	Night waking	No	12 months	No

Howlin (1984)	Stimulus fading	Case study	1	Lang delay, Autism	5.75	Settling, co-sleeping, night waking	Yes		Yes
Montgomery, Stores, & Wiggs (2004)	Routine, extinction with checking or stimulus fading, • therapist • booklet controls	Randomised controlled trial	66	Mild – Severe mixed ID and disabilities	2.3 - 8.4	Settling, night waking	Yes	6 months	Yes
Moore (2004)	Routine with social story and graduated extinction	Functional assessment, case study	1	Severe ID, autism	4	Settling, co-sleeping, night waking	Yes	None	-
O'Reilly, Lancioni, & Sigafoos (2004)	Routine, BCBC reversal using routines and fixed delivery of attention	Functional analysis, case study	1	Severe ID	5	Settling	Yes	12 months	Yes
Piazza, Fisher, & Moser (1991)	Routine, faded bedtime with response cost	Multiple baseline	3	Severe ID, Rett syndrome	8 8 4	Settling Excessive daytime sleepiness Night waking Night waking, Co-sleeping Excessive daytime sleepiness	Yes Yes Yes Yes Not reported Yes	None None None	- - -
Piazza, Fisher, & Sherer (1997)	Routine, Faded bedtime with response cost	Group design, comparison of two interventions	14	Mod-prof ID, mixed disabilities	4 - 10	Settling, night waking, early waking, shortened sleep	Yes, reduction in hours of disturbed sleep cf baseline	None	-
	Routine, Bedtime scheduling			Mod-prof ID, mixed disabilities	5 - 10	Settling, night waking, early waking, shortened sleep	No, little change in hours of disturbed sleep cf baseline	None	-
Quine (1992)	Individual programs involving routines, graded change or extinction, with positive reinforcement	Multiple baseline	25	Mixed ID and disabilities	School age	Settling, night waking, limited hours sleep	Yes (80%)	3 months	Yes (85% of original successful children)
Thackeray & Richdale (2002)	Routine Extinction	Functional assessment, delayed multiple baseline	3	Severe ID- Mod ID, DS Mild ID, ADHD, anxiety	5 5.5 10	Settling Settling, night waking, co-sleeping Settling, night waking, co-sleeping	Yes Yes, No Yes Yes, yes Yes	3 months	Yes Yes, No Yes Yes Yes
Weiskop, Matthews, & Richdale (2001)	Routine & Extinction	Functional, assessment, Case study	1	Severe ID, autism	5	Settling, co-sleeping	Yes Yes	3& 12 months	Yes Yes
Weiskop, Richdale, & Matthews (2005)	Routine & extinction *NB*: includes case in Weiskop et al. (2001)	Functional assessment, Concurrent multiple baseline, 2 groups	10	Dev delay, autism Dev delay, FRX	3.4 - 5.75 1.9 – 9.1	Settling, co-sleeping, night waking, early waking, rocking Settling, co-sleeping, night waking,, early waking	Yes No- early waking or rocking Yes, (routine only for 1 child) No – early waking	3 & 12 months 3 months	Yes, No- early waking or rocking Yes, No – early waking

Wiggs & Stores (1998)	Individual behavioural treatment	Functional assessment, Randomised design with matched control group	30	Severe – mod ID, mixed disabilities	M_T = 8.21, SD = 2.7 M_C = 10.77, SD = 3.8	Settling, night waking, co-sleeping, early waking	Yes (by parent report but not actigraphy)	2 months	Yes
Wolf, Risley, & Mees (1964)	Routine, negative consequences	Functional assessment, Case study	1	Dev delay, autism	3.5	Settling, co-sleeping	Yes	6 months	Yes

Single Case Designs

Faded Bedtime

This design involved setting a bedtime routine during baseline then appropriate sleep length, onset and wake time were established for each child using developmental norms. Faded bedtime with response cost consisted of establishing an initial bedtime when the child was likely to fall asleep quickly using baseline data for guidance. After following a bedtime routine, the child was then put to bed at this time, and woken at the pre-determined time next morning. If the child did not fall asleep within 15 minutes of bedtime the child was kept out of bed and awake for a further hour then put back to bed. If necessary this procedure continued through the night until the child fell asleep within 15 minutes. Fading from this initial bedtime then occurred by making bedtime 30 minutes earlier on nights following those where sleep was initiated within 15 minutes. If the sleep did not occur within 15 minutes bedtime was 30 minutes later the next night. Piazza, Fisher, and Moser (1991) used this approach to treat sleeplessness problems in three girls with Rett syndrome. The program was successful increasing the amount of night time sleep in two of the three girls, with a reduction in settling in one child and a reduction in the length of night waking episodes, and some reduction in their frequency in the other two girls. The two girls with excessive daytime sleepiness also showed a reduction in napping. However no follow-up data were reported, and while the intervention improved sleep the results suggest milder sleep problems still existed. As well two of the children were treated in an in-patient unit, and only one at home.

Extinction

Extinction involves ignoring or not rewarding undesired behaviours, in this case generally the child's cries or other behaviours after he has been put to bed. One advantage of extinction is that problems can often resolve within a week (e.g., Bramble, 1997) however there is also the potential disadvantage of an extinction burst where the child's behaviour becomes temporally worse before improving (e.g., Thackeray & Richdale, 2002). One of the earliest reports in the literature is that of Wolf, Risley, and Mees (1964) who addressed settling and night waking difficulties in a young boy

with autism by closing the child's door contingent upon him getting out of bed or having a tantrum in bed. The intervention was successful and progress was maintained six months later.

Individual, therapist-delivered, parent-training programs successfully addressed sleeplessness in children with an ID or Down syndrome (Thackeray & Richdale, 2002), and autism or fragile X syndrome (Weiskop, Matthews, & Richdale, 2001, Weiskop, Richdale, & Matthews, 2005). Parents were taught behavioral principles and a bedtime routine was established, followed by the implementation of extinction, with rewards the next day for good bedtime behaviour. Both studies were effective for reducing or eliminating settling problems and co-sleeping and reducing night waking, with the exception of one boy with Down syndrome whose night waking did not improve. Treatment gains were maintained at a 3-month follow-up (Thackeray & Richdale, 2002; Weiskop et al., 2001; 2005) and a 12-month follow-up for four of the children with autism (Weiskop et al, 2001; 2005). Weiskop et al.'s (2005) intervention did not improve rocking during the night or early waking, but for the oldest child with fragile X syndrome, the bedtime routine alone was successful in addressing the boy's settling problems.

For both these parent-training programs, program evaluation indicated a high level of parental satisfaction and approval. The advantages of the studies were the use of a delayed (Thackeray & Richdale, 2002) or a concurrent (Weiskop et al., 2005) multiple baseline design; functional assessment of the sleep difficulties; parent treatment goals were set, the effect of both the bedtime routine and extinction components of intervention were reported; each sleep problem was individually reported; treatment was well described and treatment manuals were used; and social validity was assessed.

Didden and colleagues reported five studies where a bedtime routine with extinction was successfully used to reduce sleeplessness in children (and one young adult) with a variety of DDs and degree of intellectual delay (Curfs, van Driel, & deMoor, 1999; Didden & Curfs, 1998; Didden, Curfs, van Driel, & deMoor, 2002; Didden, de Moor, & Curfs, 2004; Didden, de Moor, & Kruit, 1999). An individual case approach was taken and two studies (Didden et al., 2002, 2004) used a nonconcurrent multiple baseline design with a reversal design incorporated into one case (Didden et al., 2002). Across these five studies, 14 participants received an extinction program and 13 included a bedtime routine. In all cases a functional assessment was completed prior to individually implementing the intervention. The interventions were successful in reducing night-time disturbances in all reported cases except one (Didden et al., 1998) and treatment gains were maintained at either a three-month or a six-month follow-up.

Didden et al. (2004) used differential reinforcement of incompatible behaviours (DRI) with response cost using a token economy for one child with disruptive night waking. Tokens could earn desired activities and were removed for disruptive behavior. Extinction and later mild punishment (closing the bedroom door all night) were added to the treatment package. The program was eventually successful in reducing disruptive night waking, and the gains were maintained at a three-month follow-up.

Didden and colleagues' reported their data as total night-time disturbances across time, thus it is difficult to determine differential success for the individual presenting sleep difficulties; a baseline was not established for one child (Didden et al., 1998); and the effect of implementing a routine alone was not evaluated. The description of treatment was quite general in all of Didden and colleague's studies, and did not appear to involve the degree of parent training or a training manual as in Thackeray and Richdale (2002) or Weiskop et al. (2001, 2005).

Graduated Approaches

Graduated approaches to treating sleep difficulties in children include graduated extinction (where the interval before checking the child is gradually increased), ignoring with checking at fixed intervals, and stimulus fading (where parental presence is gradually faded). There are those who

believe these gradual approaches are preferable to extinction as they are less stressful for the parents and child (e.g., Lancioni, O'Reilly, & Basili, 1999).

Graduated extinction

Using a concurrent multiple baseline design, Durand, Gernott-Dott, and Mapstone (1996) used bedtime routines with graduated extinction to successfully address night waking in a child with Down syndrome and a child with 7th monosome syndrome who also co-slept, and settling problems in a child with developmental delay and a child with autism. A functional assessment was conducted, and with therapist guidance parents implemented individual programs for their child. The intervention was successful in reducing the sleep problems for the four children, including eliminating co-sleeping, with gains maintained at a six-month follow-up. According to the authors, the parents found the programs easy to implement.

Social stories were developed by Gray (1995) to accurately describe common and everyday situations that children with autism may have difficulty comprehending. They are most often used to teach socially appropriate behaviors and skills. Recently the treatment of settling and co-sleeping problems in a young boy with autism successfully incorporated a social story about bedtime (Moore, 2004). The development of the social story was preceded by a functional assessment, which indicated that attention and tangible gains were maintaining the undesirable sleep behaviors. Two favored items were chosen as key reinforcers, together with a bedtime routine. The social story, which consisted of a book of pictures showing the bedtime routine and its consequences, was read once before bedtime. The presence of mother at settling and any night waking were addressed using graduated extinction. Positive changes were seen after the first night, and were maintained at 28 days when the graduated extinction program was modified. Mother approved of the program and felt that the changes had generalized to improvements in daytime behavior. This case study illustrates a different approach to teaching a child bedtime expectations as part of a behavior management program and may be particularly useful when a child has significant communication problems. However little sleep data were presented to support the changes reported, with the study focussing on the social story.

Fixed interval attention

O'Reilly, Lancioni, and Sigafoos (2004) reported on a single case of a 5-year-old girl with a settling problem where a functional analysis was conducted which determined that the mother's presence was maintaining the settling problem. A bedtime routine was put in place, and then mother provided attention for 20 s on fixed 5 min schedule until the girl was asleep. A BCBC reversal design illustrated that fixed attention, but not the routine, was successful in reducing the number of times the child left her bedroom and in improving in her sleep latency. Improvements in settling behavior were maintained at a 12-month follow-up. The major strengths of this study were its unique use of functional analysis and the reversal design that clearly showed the effectiveness of fixed attention in addressing the settling problems.

Stimulus fading

A single case study using a stimulus fading procedure was successful in addressing sleep problems in a young boy with autism (Howlin, 1984). This boy would not sleep without his mother, needing her presence to fall asleep and if he woke during the night. After a two-week baseline period, mother introduced an air-mattress into his bedroom, and began sleeping on the mattress next to his bed. Over a period of eight weeks the mattress was gradually moved further from the boy's bed, to the door and finally into the parents' bedroom where mother returned to her own bed. The intervention was successful in eliminating settling and co-sleeping, and reducing night waking and gains were maintained at a three- and six-month follow-up. In addition the mother had ceased to take anti-depressants, which had initially been prescribed due to the stresses of the child's behavior, and the boy would now have a babysitter. The intervention was simple and clearly described, but no daily or weekly data were presented.

A single case series, using stimulus fading together with a bedtime routine and relaxation was conducted by Hewitt (1985) to address settling and night waking. Ten children with severe ID and mostly Down syndrome (7 cases) completed individually tailored and supported interventions. Functional assessments were conducted and after the bedtime routine, which included relaxation activities, parents gradually distanced themselves from the child. Night waking was treated in a similar fashion, with minimal engagement with the child. The intervention was successful with seven of the children, and in one unsuccessful case, the cause of the problem appeared to be epilepsy. Gains were maintained at follow-up, which was at 12 months in most cases. Even when there had been a relapse prior to the 12 months, most parents were able to successfully reinstate good sleep by follow-up. This was a therapist directed program and the paper gives clear written summaries for each child at baseline, the end of treatment and follow-up but though daily data were recorded, no data are presented. The program is only briefly described, though parents were given individual written programs; as well parents decided when to stop recording data based on their satisfaction with their child's sleep. Thus the level of experimenter control does not seem to be high.

Desensitisation with differential reinforcement of other behavior

In their 1998 paper Didden et al. had one participant for whom anxiety was hypothesised to be maintaining settling problems. A desensitisation program, with differential reinforcement of appropriate bedtime behavior was used. The boy was gradually accustomed to his mother's absence and quiet behavior was rewarded with an edible reinforcer. The program was successful and gains maintained at six-months follow up. However there were no baseline data for this case.

Sleep restriction

Two recent additions to the literature involved the use of a sleep restriction program (Durand & Christodulou, 2004; Christodulu & Durand, 2004) in a concurrent multiple baseline design. In the former study a young girl with autism and settling difficulties, night waking, and co-sleeping and second girl with developmental delay and similar sleep problems were dealt with by restricting their sleep to 90% of normal sleep length. An initial assessment of the sleep problems and baseline data were collected, initial bedtime was moved to a later time, and a fixed wake time was employed. When the initial sleep disturbances were reduced bedtime was faded back to an age-appropriate time. In the latter study a similar program, with the addition of a bedtime routine, was used to address sleeplessness in four developmentally delayed children. A reversal phase was incorporated for one child and actigraphy was used to confirm parental diaries in another child. The intervention was successful for all children, with one child's sleep difficulties being resolved with the bedtime routine alone. Gains were maintained at a follow-up, one month post-intervention.

Parent satisfaction data indicated that parents approved of the sleep restriction intervention and considered their child's sleep problems to have significantly improved. As well the authors claim that its major advantage is that it reduces or eliminates the likelihood of an extinction burst or temporary increase in difficult sleep behaviors, often seen in extinction or graduated extinction procedures.

Group Designs

Extinction

Using a group approach to data analysis, Bramble (1996, 1997) reported on 15 children with severe ID and life-long sleep difficulties, including settling, night waking and co-sleeping. Each family received standard advice with only minimal individual tailoring. Parents were given a single face-to-face session (with subsequent phone contact support as required) during which they were advised to set and stick to a regular bedtime for their child; implement a calming bedtime routine and set the mood for sleep leading up to bedtime (i.e., no play, being quiet and calm); settle their child in the bedroom rapidly and then leave the room; thereafter ignore their child (unless unwell) only intervening to ask their child to return to the bedroom, if they came out of it or, if necessary, physically return their child to the room, as often as necessary. Good night time behavior was also to be rewarded. Group data of parents' ratings of daily sleep problem severity were reported. Extinction was successful in reducing the presenting sleep problems for the majority of children, with

improvements generally maintained at 4- and 18-month follow-ups. Parental satisfaction with the both the treatment and the treatment outcome was high.

Major advantages of this study were the consideration of parental satisfaction and the 18-month follow-up. The intervention program was also quite clearly described. However the use of group data, and missing data make it impossible to determine the individual effectiveness for specific presenting problems for each child. Also there was no comparison group.

Graduated Extinction

In the first randomised controlled, cross-over design in this area, Montgomery, Stores, and Wiggs (2004) compared two behavioral interventions for sleep problems in 66 children with an ID or other DDs. Presenting sleep problems included settling and night waking. The intervention involved either a therapist-guided intervention using a written manual or giving parents the written manual. The manual covered information about children's sleep, behavioral principles and sleep intervention, and using bedtime routines and graded extinction, with rewards next day for good bedtime behavior. Group data concerning sleep problem severity were reported and the two intervention approaches were compared with a wait-list control, which then received one or other of the interventions using a cross-over design. The interventions were equally successful with participants in each group showing significant improvement in their sleep. No improvement occurred in the control group until they received intervention and gains were maintained at six-month follow-up.

About 70% of parents approved of the intervention techniques, and the intervention appeared successful for the majority of participants. This program is unique as it represents the only randomised controlled trial that compares two interventions with a control condition, greatly strengthening the confidence one can have in its outcomes. It also includes a written manual. However individual intervention effects on settling and night waking were not reported and it would have been interesting to know about participants for whom the intervention was not successful. The latter may provide guidance regarding those who will require a therapist-guided parent training program and those for whom written information will be sufficient to resolve any child sleeplessness difficulties.

Individually Tailored Behavioral Interventions

In 1992 Quine reported on a series of 25 school-aged children with an ID and a variety of sleep difficulties including settling, night waking and limited hours of sleep. Programs were individually tailored for each child and involved bedtime routines, extinction, or graded change and positive reinforcement. Parents were assisted to implement their child's program by a Health Visitor. Group data were reported and the behavioral intervention was successful for 80% of the children with success being maintained for 85% of this group at a three-month follow-up. There was no comparison group.

An individual approach to intervention was also used by Wiggs and Stores (1998). Parents of 30 children with an ID and various DDs were randomly assigned on the basis of their school to either a treatment or a control group; groups were matched on the duration of the sleep problem. A therapist-guided program included home visits and a functional assessment; treatment goals; behaviors related to sleep; the presentation of several options for sleep intervention including bedtime routines, extinction, graded extinction, stimulus fading, and positive reinforcement; and an individual written intervention program. A composite sleep index score indicated the severity of the sleep problems and actigraphy was used to monitor sleep at pre-and post-intervention and at the two-month follow-up. The composite sleep index scores indicated that compared to the control group, sleep had improved after intervention and the gains were maintained at follow-up. However, this was not clearly supported by the actigraphy data as improvements occurred in both the treatment and control groups from pre- to post-intervention, and this was generally maintained at follow-up. Objective improvements in the mothers' sleep were only seen in the treatment group.

Wiggs and Stores (1998) suggested that the difference between the subjective and objective reports of the children's sleep may be because when they were awake children no longer disturbed their parents, which benefited the mothers' night time sleep. This study has two major strengths, comparison with a control group and objective measurement of sleep parameters in both the child and mother. However, no conclusions can be drawn about the effects on any improvements in settling behavior or sleep latency as data were not specifically reported for these parameters. The objective improvements in the control group's sleep suggest that there needs to be a careful examination of parent and child behaviors surrounding sleep problems in children with DDs.

Faded Bedtime with Response Cost Versus Sleep Scheduling.

A comparison of faded bedtime with response cost and bedtime scheduling to treat multiple sleep problems (settling, night waking, early waking, shortened night sleep) in 14 children with severe ID and a range of DDs was conducted by Piazza, Fisher, and Sherer (1997). Appropriate sleep length, onset and wake time were established for each child using developmental norms. The faded bedtime with response cost intervention was the same as that reported above for Piazza et al. (1991). In the bedtime scheduling condition a bedtime routine was established and developmentally appropriate bedtimes and wake times were selected and adhered to consistently. Seven children completed each intervention. Faded bedtime with response cost was effective in addressing sleep problems as indicated by a significant reduction in hours of disturbed sleep, with all children showing improvements which, in most cases were considerable. Bedtime scheduling produced little change in hours of disturbed sleep and had only a modest effect on some children. No follow-up was conducted and there was no control group.

Impact of Sleep Intervention on Daytime Behavior

Wiggs and Stores (1999) reported on behavior change as a result of their sleep intervention package (Wiggs & Stores, 1998), and found that at post-intervention and follow-up some behaviors improved in severity, but not frequency, and that improvements occurred in the control group as well. The effects of intervention on both mothers and father was assessed (Wiggs & Stores, 2001), with mothers reporting increased satisfaction with their own sleep, their child's sleep and their ability to cope with the child's sleep, particularly in the treatment group. More modest but similar improvements were seen for fathers in the treatment group and, to some extent, the control group. As mentioned, the mothers' objective total sleep duration also increased from pre- to post-intervention compared with the control group.

Bramble (1997), Thackeray and Richdale (2002) and Weiskop (2001) also assessed generalisation to daytime behavior resulting from their sleep intervention packages (Bramble, 1996; Thackeray & Richdale, 2002; Weiskop et al., 2001, 2005). Bramble found that daytime behavior problems as measured by a questionnaire improved from pre-treatment to the 4-month follow-up, and Thackeray and Richdale also found some evidence for behavior improvement using a questionnaire. However using both a questionnaire and video analysis of mother-child interactions Weiskop found no meaningful improvements in daytime behavior. Similarly, Thackeray and Richdale also measured specific target behaviors and conducted classroom behavior observations and found that support for improvements in daytime behavior was equivocal. In addition, whilst Bramble reported that maternal stress and sleep improved as a result of treating the sleep problems, but Weiskop found no meaningful changes in maternal stress.

Differences in child behavior change and parent responses to these sleep interventions may be due to small sample size; differences in the measures used to assess behavior change and parent stress; and child and family factors such as the type and severity of the child's disability, and social supports available to families. This emphasises the difficulties in evaluating any generalisation of sleep improvements to other child behaviors and to family members.

Conclusions Regarding the Treatment of Sleeplessness

It is difficult to classify the above studies with regard to the criteria for empirically supported psychological interventions (Chambless & Ollendick, 2001; Lonigan et al., 1998) as the populations of developmentally delayed children included are heterogeneous, as are behavioral approaches used. There is also considerable variation in the studies' designs, their design quality, and the choice of outcome measures. However, all treatments were effective for reducing or even eliminating most presenting sleeplessness problems for almost all the participants. The majority of studies incorporated a bedtime routine (regular bedtime activities with a fixed bedtime) into their treatment package and extinction was the most commonly used approach. Parents kept daily sleep diaries, and some form of functional assessment of the sleep behaviors was also conducted. One study incorporated a more demanding functional analysis of the behavior(s) maintaining the presenting sleep problem (O'Reilly et al., 2004).

This array of studies indicates that sleeplessness in children with DDs can be successfully addressed using a variety of behavioral approaches, tailored to the individual needs of the child and their family. Extinction has the most empirical support as there are now at least eight case studies ranging from single subjects to multiple baseline designs and one group study that have reported good outcomes. However, based on the studies reviewed here, it does not meet criteria for a well-established treatment for children with DDs. Studies have predominantly emanated from two investigator groups; it is not clear that all studies used treatment manuals; and while all studies incorporated a bedtime routine within their design, few compared this intervention component alone with extinction, or compared extinction with any other form of intervention. Extinction clearly meets criteria for a probably efficacious treatment (Chambless & Ollendick, 2001; Lonigan et al., 1998) for children with DDs provided one accepts that it has been used across children with a heterogeneous set of conditions. As well the social validity data reported indicate that parents approve of extinction and are satisfied with its outcomes. The large randomised controlled trial of Montgomery et al. (2004), which used graduated extinction, also meets Chambless and Ollendick's criteria for a probably efficacious treatment. While all of the other reports are encouraging, there are insufficient numbers of studies, generally a lack of control groups or comparison interventions, and it is unclear that all investigators used treatment manuals. Thus none of the other approaches reviewed currently meets criteria for a probably efficacious treatment and must still be considered experimental in this population of children.

Sleep-Wake Rhythm Disorders

Sleep-wake rhythm disorders refer to problems with the timing of sleep within the 24-hour day. Sleep may occur too early (phase advanced), or too late (phase delayed); it may be fragmented occurring irregularly throughout the 24 hours; or be free-running, that is dissociated from regular time cues with sleep onset gradually delaying over successive nights thus moving the sleep period around the 24 hour clock over a period of days or weeks. The light-dark cycle is the most important zeitgeber or time-cue for resetting the sleep/wake rhythm to the 24-hour day, with other environmental cues such as regular social interactions and meals acting as secondary zeitgebers. The most common causes of a phase advance or delay in the sleep / wake rhythm are those associated with jet lag, that is the when the sleep-wake rhythm is desynchronised relative to local time due to crossing several time zones in a short time period during flights; and shift work. Phase delays in the sleep-wake rhythm are also common in adolescents who, due to the demands of study and their social lives, often begin going to bed in the early hours of the morning, and are thus difficult to wake up and tired when they must later get up for school, or who sleep until afternoon when the opportunity arises (e.g., on weekends). Fragmented and sometimes free-running sleep/wake rhythms are most commonly found in those with significant visual impairment (lack of light perception) (Sack, Lewy, Blood, Keith, &

Nakagawa, 1992) Children with significant brain impairment are also at high risk for circadian sleep disturbances (Okawa & Sasaki, 1987).

Treatment of Sleep/Wake Rhythm Disorders

Approaches to treating sleep/wake rhythm problems generally involve resetting the circadian clock using chronobiological approaches. These include bright light treatment, or altering sleep and wake times behaviorally (chronotherapy), generally by successively phase delaying sleep until the desired bedtime is reached, or forced waking; or prescribing the neurohormone melatonin to be taken prior to sleep onset. Strong social cues (i.e., regular meals and daily routine and a regular bedtime routine) that provide additional zeitgebers are also put in place. For a discussion of melatonin for the treatment of sleep problems in children with DDs the reader is referred to Phillips and Appleton (2004).

Bright light therapy involves either morning light exposure to cause a phase advance in the sleep-wake rhythm, or evening bright light exposure, which results in a phase delay. It should be noted that 'morning' and 'evening' are relative to the individual's own body clock time and this will not necessarily correspond with the actual clock-time. Guilleminault et al. (1993) used this approach, together with strict daytime and bedtime routines to successfully treat intractable sleep problems in 5 of 14 children with severe DDs, with treatment gains being maintained for several years. The authors concluded that bright light therapy may be useful when other treatments have failed. Okawa et al. (1987) used chronobiological approaches with four congenitally blind girls (age 4-12 years) with a moderate to severe ID and a fragmented (1 case) or free-running sleep-wake cycle (3 cases). Bright light therapy was unsuccessful for one child, and the other three children were treated using forced waking and regular daytime schedules and night routines that acted as zeitgebers. The latter approach was successful for one case and the mother continued to enforce the routines at home. One child improved when epileptic seizures were better controlled and the third case did not improve. The sleep problems were thought most likely related to the lack of social cues to entrain the rhythms and the type and site of brain damage associated with the girls' conditions. While the case descriptions for these girls were quite detailed, information about the sleep interventions and their success was quite limited.

Piazza et al. (1998) successfully treated an 8-year-old girl with severe ID and autism and a fragmented sleep-wake pattern and reduced total sleep using chronotherapy together with regular daytime activities. After baseline data were collected, an appropriate time when the girl was likely to be asleep was selected as the initial bedtime (3.30 am). The child's bedtime was successively and systematically phased delayed until the desired bedtime was reached. Phase delay is more appropriate than phase advance because of our natural tendency to be able to fall asleep later than normal, rather than earlier. An increase in the length of night time sleep, improvements in settling and sleep latency, reductions in night waking and a more appropriate sleep pattern resulted. The gains were maintained at home at 4-month follow-up.

Thus the treatment of severe sleep-wake rhythm disturbances in children with DDs using chronobiological approaches is still in its infancy. Bright light treatment (which is not strictly a behavioral approach) appears successful in a minority of reported cases, and while it has potential there are too few cases reported where the sleep-wake rhythm has been altered behaviorally, using forced waking or phase delay. Both Piazza et al. (1998) and Okawa et al. (1987) treated their clients in a hospital setting over a period of weeks and it would likely be difficult for parents to conduct similar interventions in the home, at least not without extensive clinical support. The case study by Piazza et al. however appears most promising and the approach deserves further attention by sleep researchers working with children with DDs.

Parasomnias

The parasomnias are the category of sleep disorders referring to episodes of behavior, experiences and/or physiological events, which occur in relation to sleep. They are typically divided into four categories, according to the stage of sleep during which they occur: (a) sleep-wake transition disorders which occur pre-sleep and during sleep onset (e.g., rhythmic movement disorders, restless legs syndrome), (b) parasomnias associated with Rapid Eye Movement (REM) sleep (e.g., nightmares), (c) arousal disorders which involve a sudden, partial arousal to light Non REM sleep from deep Non REM sleep (e.g., sleep walking, sleep terrors); and (d) those parasomnias not consistently related to any particular stage of sleep (e.g., teethgrinding).

Research documenting the prevalence and treatment of parasomnias in children with DDs has been limited. There have been some studies suggesting that parasomnias might be more common in children with DDs (Schreck & Mulick, 2000) whereas others suggest the opposite (Wiggs & Stores, 1996b; Wiggs & Stores, 2004). Specific parasomnias have been noted to occur in children with particular underlying conditions, for example, REM sleep behavior disorder in a series of children with autism (Thirumalai, Shubin, & Robinson, 2002), periodic limb movements in children with attention deficit hyperactivity disorder (Chervin et al., 2002) and Williams syndrome (Arens et al., 1998). Some parasomnias have also been documented to occur in association with other sleep disorders e.g., arousal disorders and enuresis in children with sleep related breathing difficulties (Guilleminault & Khramtsov 2001).

Many of the most common parasomnias of childhood are typically benign and/or developmental phenomena which are likely to remit spontaneously in time. Other parasomnias, if severe, persistent or compromising sleep and/or physical or psychological well-being will require treatment. Appropriate treatment, of course, will vary depending on the nature of the presenting parasomnia but there have been reports of behavioral interventions being used successfully to treat TD children with sleepwalking, sleep terrors, nightmares and rhythmic movements disorders (see Kuhn & Elliott, 2003 for a review). There certainly remains a need for much more work in this area. For children with DDs the literature is even more limited, with reports of behavioral interventions confined to the treatment of sleep terrors and, to some extent, rhythmic movement disorders.

Sleep Terrors

Durand (2002) describes the use of scheduled wakings to treat sleep terrors in three children with autism, aged between 3 and 7 years. The parents kept sleep diary records of their child's sleep patterns for 25 weeks and from the baseline data the average time of occurrence of their child's sleep terror could be calculated (sleep terrors typically being predictable in their timing and occurring within a couple of hours after sleep onset, to coincide with the child being in deep Non REM sleep). At the start of the intervention phase parents were instructed to wake their child, with a light touch, just to the point when their child opened their eyes, and then let them fall back to sleep again. They continued doing this until the child had seven nights without a sleep terror at which point they did no scheduled waking for one night, then six nights of scheduled waking. If no sleep terrors occurred for seven nights they then skipped two nights and performed scheduled waking for the remaining five nights of the week. Scheduled wakings were gradually reduced in this manner. A multiple baseline across subjects design was used, with weekly frequency data clearly documenting the immediate effects of treatment at reducing the frequency of episodes to zero or less than one per week, with the effects being maintained at 12 months follow-up at which time all children were reported to have zero episodes. The authors speculate on the possible mechanisms underlying successful treatment. It may be that, the waking caused a reorganization of the child's deep sleep which eliminated the sleep terrors or that the procedure conditioned the children to self-arouse. The study did not include polysomnography so it was not possible to explore these hypotheses. Of further note was that the sleep duration of two of the children was increased by the intervention. As sleep deprivation may provoke sleep terrors in susceptible children (Mindell & Owens, 2003), this increase in sleep may be a contributory factor to the treatment's success.

Helpfully, the study included important checks on the accuracy of the parents' reports and treatment compliance as well as documenting parental satisfaction with the treatment and the outcome. Given the paucity of knowledge surrounding the presentation of, and factors associated with, sleep terrors in children with DDs it is a shame that the clinical descriptions of the children and their episodes are limited and the diagnoses of sleep terrors are based upon the opinion of a single clinician, without any polysomnographic data to support the diagnosis. This is of note because, of course, scheduled wakings has also be used successfully with TD children as a treatment for nightwakings which are not due to an arousal disorder (Rickert & Johnson, 1988). Consistent with the diagnosis of sleep terrors was the predictable timing of the episodes, that they began with a scream, that the children were distressed and physically agitated whilst non-responsive during the episodes and that sleep-walking (another arousal disorder which can co-occur) co-existed in two children. A somewhat unusual feature in one child was that the episodes began when the child was 3 months old. Information about the duration of episodes, family history and how these night time episodes differed behaviorally (or otherwise) from the other 'nightwakings' that the children were reported to suffer from would have been useful to increase clinical understanding.

Rhythmic Movement Disorders

Rhythmic movement disorders are stereotyped, rhythmic movements of the head, neck, limbs and/or trunk, which occur in association with sleep (either during pre-sleep wakefulness/drowsiness or during sleep itself). There are some definitional problems with the literature concerning rhythmic movement disorders in people with DDs, partly because rhythmic movements are more likely to also occur during the day (perhaps, but not necessarily, for different reasons) and also because they might be confused with generalised self-injurious behavior (SIB). Many of the reports of behavioral interventions for head banging are actually treatment of SIB, variably occurring in relation to sleep (e.g., DeCatanzano & Baldwin, 1978; Prochaska, Smith, Marzilli, Colby, & Donovan, 1974). Two studies deserve mention in this review because they report a connection between the behavior and sleep and because the behavior is of a sort which could be a rhythmic movement disorder (i.e., head banging and not other forms of SIB such as striking fists into the eyes) although whether the children's rhythmic movements meet the criteria for rhythmic movement disorder as stated in the International Classification of Sleep Disorders (ASDA, 2001) is questionable.

Weiher and Harman (1975) describe the successful use of omission training (i.e., positively reinforcing the non-occurrence of behavior) to treat head banging in a 14-year-old child with Down syndrome and severe ID. The child banged his head during the day against hard surfaces and at night against the bed frame. Therapy was delivered during the daytime. Using a controlled reversal design resulted in a significant reduction in head banging (to nearly zero) and an increase in prosocial behaviors both of which were maintained during the reversal condition when baseline conditions were re-instated. The reinforcement consisted of providing small amounts of apple sauce if no head banging occurred during gradually extending pre-set time intervals.

DeLeon, Fisher, and Marhefka (2004) also used a behavioral approach in a four year old boy with autism to address SIB (including head banging) which occurred with greatest frequency following awakenings. The particularly novel aspect of this study was that the intervention was directed towards the child's sleep pattern rather than at the target behavior itself; a form of sleep scheduling was used to consolidate sleep and minimise nightwakes. The child was assigned a scheduled bedtime (initially set late to permit only the hours of uninterrupted sleep he achieved during baseline) and wake up time, and daytime sleep was not permitted. Bedtime was gradually faded earlier in 30-15 minute increments contingent upon him falling asleep within 30 minutes and having no nightwakings or early wakings of more than 30 minutes. The treatment reduced both his night waking episodes and the number and duration of SIB episodes. The study's merits include the 24-hour observation and documenting of both sleep and SIB, with reports of hourly frequency data and some checks for inter-observer agreement. The authors acknowledge the limitations of the AB design and the fact that the operant mechanism involved in the relation between wakings and SIB could not be

determined. However, they discuss interesting possible mechanisms, which suggest future potential research areas.

Encouraging though these preliminary reports are, there is clearly a need for controlled studies of behavioral interventions for well-defined parasomnias that meet standardized diagnostic criteria before conclusions can be drawn about the efficacy of behavioral therapy in the treatment of parasomnias in children with DDs.

Excessive Time in Bed

Excessive sleepiness is usually either due to conditions, which produce an increased need for sleep (e.g., narcolepsy, Kleine Levin syndrome) or it arises as a result of impaired nocturnal sleep quality and/or quantity. Sleeplessness problems, sleep-wake cycle disorders and various parasomnias may all, if severe enough, compromise the amount of sleep obtained or disturb sleep sufficiently to result in daytime sleepiness. Sleep related breathing disorders, which result in arousals, are a common cause of impaired sleep quality which feature prominently in persons with DDs. Approaches to the management of excessive sleepiness will vary according to the disorder underlying the sleepiness (e.g., surgical intervention or ventilation for sleep related breathing disorders, primarily pharmacological symptomatic relief for narcolepsy). Documented behavioral approaches to the treatment of sleeplessness, sleep-wake cycle disorders and parasomnias in children with DD have been discussed earlier in this paper.

An associated problem, which has been reported in children (and adults) with DDs, is that of excessive time in bed. This may be mistakenly construed as excessive sleepiness or even present as sleeplessness, depending on the child's behavior whilst in bed. Espie (1992) also makes the point that putting the child to bed may be a parental coping response, in the hope that this will provide a form of respite. This may not actually be to the benefit of the child or their parents.

Treatment of Excessive Time in Bed

Espie (1992) describes using optimal sleep-wake scheduling to consolidate sleep and improve daytime functioning and behavior in a 16-year-old girl with profound ID. This intervention is a form of stimulus control technique, designed to consolidate sleep and to ensure that a greater proportion of time spent in bed is actually spent asleep. This is achieved by setting a bedtime which is gradually moved to co-ordinate with normal sleep onset time, while total time in bed is restricted by setting a wake-up time that only allows enough time for an individual's total nightly sleep duration (calculated from baseline records). Napping is prohibited and these measures result in an initial degree of sleep deprivation, which makes it more likely that the individual will fall asleep quickly and have more consolidated sleep. Once more than 90% of the time in bed is spent asleep the bedtime is gradually brought forward, as appropriate, to ensure that sleep duration is maximized while time in bed spent awake is minimised.

In this uncontrolled report Espie (1992) provides weekly frequency data of sleep duration, restedness after sleep and daytime behavior over a 22-week period while these changes to the sleep schedule are gradually implemented. He notes the possible confounding effect of concomitant Haloperidol withdrawal but suggests that the data indicate a possible relationship between improved sleep pattern and improved daytime behavior (measured by daily logs and questionnaires completed by different informants). Espie and Wilson (1993) provide a further three case reports of the use of optimal sleep-wake scheduling with three, 12-18 year old males with mild/moderate ID. Mean data for the final baseline week and final post-treatment week are given indicating that sleep latency reduced for two cases, night time wakings reduced for two cases, night-sleep duration increased for one case and the clock-time at which the person fell asleep became more appropriate for one case. Where aspects of daytime functioning were recorded (two cases) improvements were noted.

Although these reports do not meet criteria for a probably efficacious treatment they do highlight a strategy, which should be subjected to further empirical study, especially as it has the potential to be used preventively to improve sleep-wake patterns in a population for which disruption of the sleep-wake pattern is commonplace.

Conclusions

The treatment of sleep disorders or problems in children with an ID or DDs is in its infancy. The state of research is such that only extinction and graduated extinction (both with bedtime routines) meet criteria for probably efficacious treatments for sleeplessness in these children. Other behavioral treatment studies for sleeplessness, and behavioral treatments for other sleep problems or disorders are either non-existent or consist of single studies of one or a few cases, making them experimental at best. Thus the state of our knowledge concerning the most effective and efficacious behavioral treatments for sleep problems in these children is even worse than that reported for TD children (see Kuhn & Elliot, 2003).

However, the preliminary work suggests that many common sleep problems are often behaviorally maintained or have a behavioral component even when there is an underlying medical or developmental condition. Psychologists and others with a sound knowledge of behavior therapy, DDs and sleep are in the best position to further our knowledge by conducting soundly designed, controlled intervention studies to determine the relative effectiveness and efficacy of behavioral approaches and to further understanding about how these may be variously affected by the type of sleep disorder and psycho-social/medical factors in individual children and their families. For example, factors that are rewarding or punishing in the context of an extinction program for a child with autism, might be expected to be quite different from those for a child with Down syndrome, due to the differing intrinsic behavioral, communication and social factors underlying each condition. Controlled studies with discrete samples (both in terms of underlying condition and the type of sleep disorder) utilizing both subjective and objective measurement of sleep will be most helpful.

The role of pharmacological intervention in the treatment of sleep disorders in children with DDs is not clear; basic safety and efficacy data are lacking and, where sleep disorders are being maintained by a behavioral element, behavioral interventions are likely to be of longer-term benefit. Nevertheless, the potential of combining pharmacological and behavioral interventions needs experimental exploration, perhaps especially for use with 'hard-to-treat' children, as this combined approach has been used successfully in TD samples (France, Blampied, & Wilkinson, 1991).

In addition to the need for quality research there is also a need to increase basic awareness and education about sleep, its disorders and treatment possibilities amongst clinicians, parents and other professionals (e.g., teachers) involved in the care of children with DDs. Established sleep problems need to be recognized and the appropriate services and personnel be available to accurately assess, diagnose and treat as needed. Ideally, services would be 'phased' so that many common behavioral sleep disorders could be identified and treated at the primary and secondary health-care levels with referral to specialised sleep disorders services, which are limited, reserved for cases where specialized assessment/management is needed.

Finally, given the prevalence of sleep disorders in children with DDs the importance of health-promotion and preventive approaches should also be emphasised. Instructing parents in ways to encourage the development of good sleep patterns may help prevent some of the long-standing and severe sleep disorders and their associated negative effects for the child and their family.

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